REVIEW ARTICLE

Oral Pathology

Frequency of odontogenic cysts and tumors: a systematic review

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Abstract

A systematic review of the literature from 1993 to 2011 was undertaken examining frequency data of the most common odontogenic cysts and tumors. Seven inclusion criteria were met for the paper to be incorporated. In the preliminary search 5231 papers were identified, of these 26 papers met the inclusion criteria. There were 18 297 odontogenic cysts reported. Of these there were 9982 (54.6%) radicular cysts, 3772 (20.6%) dentigerous cysts and 2145 (11.7%) keratocystic odontogenic tumors. With the reclassification of keratocystic odontogenic tumor in 2005 as an odontogenic tumor, there were 8129 odontogenic tumors reported with 3001 (36.9%) ameloblastomas, 1163 (14.3%) keratocystic odontogenic tumors, 533 (6.5%) odontogenic myxomas, 337 (4.1%) adenomatoid odontogenic tumors and 127 (1.6%) ameloblastic fibromas. This systematic review found that odontogenic cysts are 2.25 times more frequent than odontogenic tumors. The most frequent odontogenic cyst and tumor were the radicular cyst and ameloblastoma respectively.

Introduction

Epidemiological data for odontogenic cysts and tumors have been reported extensively in the literature, with an emphasis on the more destructive lesions such as the keratocystic odontogenic tumor (KCOT) and ameloblastoma. Various statistical terms have been used by authors when discussing epidemiological data. Frequency is the number of times a case occurs in a given period, whereas prevalence is the total number of cases of a disease in a given population at a specific time. Incidence is the number of new cases of a specific disease occurring during a certain period in a population.

Odontogenic lesions can be divided discretely into cysts (an epithelial lined pathological cavity) and tumors (a solid tissue mass, not necessarily neoplastic).[1] Intrabony lesions of the jaws are identified most commonly on radiographic imaging frequently as an incidental finding.[2]

A radicular cyst is due to a long standing inflammatory process in the bone of the surrounding root apex with the proliferation of the epithelial rests of Malassez (residual epithelial cells in the periodontal ligament).[3]

Dentigerous cysts are developmental in origin, with fluid expanding the dental follicle peri-coronally.[4] The KCOT arises from the dental lamina and other sources of odontogenic epithelium.[5,6] This lesion tends to be more aggressive in its growth pattern, providing a higher recurrence rate than other odontogenic pathoses.[7–9]

Ameloblastomas can be derived from any odontogenic epithelium ranging from the dental lamina (pre-odontogenesis), to reduced enamel epithelium (post-odontogenesis), to epithelial rests of Malassez and Serres (post-eruption), and possibly the basal layer of the overlying epithelium (the primitive source of dental lamina during embryogenesis and pre-odontogenesis).[10–12] Understandably then ameloblastomas can have a varied radiographic appearance.

Odontogenic myxoma is an intraosseous neoplasm consisting of myxomatous fibrous extracellular matrix originating from mesenchymal remnants.[13] Adenomatoid odontogenic tumors arise from the dental lamina in the gubernacular cord of developing permanent teeth.[14] Ameloblastic fibromas are similar in origin to ameloblastomas, being derived from the enamel organ or dental lamina, except there is a lack of dental hard tissue in the specimen.[13]

Frequency of odontogenic cysts and tumors is important, as patients deserve a realistic opinion of likely

diagnoses when radiographic lucencies are an incidental finding on panoramic radiographs such that an emphasis is not placed on uncommon, albeit aggressive lesions.

The aim of this study was to systematically review all papers between 1993 and 2011 that provided frequency data for odontogenic cysts and tumors. When combining the data, an overall frequency for the various odontogenic cysts and tumors was calculated. This is important for all healthcare professionals in providing a global frequency for their patients. As most of the papers presented retrospective data, 1993 was seen as an appropriate starting point enabling the authors to provide up-to-date frequency values.

Methods

A systematic review was conducted using the databases CENTRAL, MEDLINE, and EMBASE. An electronic search for papers published in English that presented data on odontogenic cysts and tumors (with appropriate synonyms) was performed. For papers to be included in the systematic review, seven inclusion criteria had to be met (Table 1).

Papers describing odontogenic cysts had to present data on radicular cyst, dentigerous cyst and KCOT. Papers describing odontogenic tumors had to present data on ameloblastoma, odontogenic myxoma, adenomatoid odontogenic tumor, and ameloblastic fibroma. Papers had to state which World Health Organization (WHO) classification system they used and describe the location from where the data were collected. The papers had to report the total number of lesions collected over a specific time frame, along with the individual numbers of the various lesions. Papers were excluded if they did not meet all of the inclusion criteria.

Table 1. Inclusion criteria

1	The paper must be full text and written in English
2	The paper must describe frequency or prevalence of odontogenic cysts or tumors
3	Papers must state which WHO classification system was used
4	Papers describing odontogenic cysts using the 1992 WHO classification must report data on radicular cyst, dentigerous cyst and KCOT
5	Papers describing odontogenic tumors must report data on ameloblastoma, odontogenic myxoma, adenomatoid odontogenic tumor and ameloblastic fibroma
6	Papers describing odontogenic tumors using the 2005 WHO classification must report data on ameloblastoma, KCOT, odontogenic myxoma, adenomatoid odontogenic tumor and ameloblastic fibroma
7	Papers had to report the total number of lesions collected over a specific time frame, along with the individual numbers of the various lesions

KCOT, keratocystic odontogenic tumor.

Titles and abstracts were perused by two reviewers (NJ, MB) for papers describing frequency of odontogenic cysts or tumors. Articles satisfying the inclusion criteria were retrieved in full text for further analysis.

Results

There were 5231 papers identified in the preliminary database search. Of these there were 10 papers that met the inclusion criteria that described odontogenic cysts. There were 12 papers that met the inclusion criteria that described odontogenic tumors. With the change of the WHO classification describing KCOT as a tumor instead of a cyst in 2005,[13] there was a subset of four papers that dealt with KCOT under the odontogenic tumor classification. All selected papers dealt with populations ranging from the first to eighth decades.

To better appreciate the global frequency of the various pathoses further data analysis was undertaken. The total number of cysts or tumors reported in each paper was combined. The total number of each individual pathological lesion was then divided by the total number of cysts or tumors. This provided an overall frequency for the individual pathoses and was seen as the most appropriate analysis of the data available.

When reviewing the papers that described odontogenic cysts (Table 2), the frequency of radicular cysts spanned 37.9%[15] to 84.5%[16] dentigerous cysts ranged between 11.4%[16] and 35.5%,[17] and KCOT ranged between 1.3%[16] and 21.5%.[18] When combining the data from all of the papers in Table 2, there were 18 297 cysts reported. Of these there were 9982 radicular cysts with a frequency of 54.6%, 3772 dentigerous cysts with a frequency of 20.6% and 2145 KCOTs with a frequency of 11.7%.

In 2005 KCOT was reclassified by the WHO as a tumor rather than a cyst.[13] As such, papers published prior to 2005 refer to KCOT as a cyst, including this entity in their cyst pathology numbers (Table 2). Many papers even after 2005 still incorporated KCOT into their cyst data,[16,19,20] with some stating[16,20] they used the 1992 WHO classification system.[21]

The frequencies of selected odontogenic tumors are presented in Tables 3 and 4. Table 3 represents the data published prior to the reclassification of KCOT and Table 4 represents the data after the reclassification. There were 8129 odontogenic tumors, which were derived from the data in Tables 3 and 4.

With regards to the data presented in Table 3, the frequency of ameloblastoma ranged from 13.5%[22] to 80.1%,[23] odontogenic myxoma 2.2%[24] to 17.7%,[25] adenomatoid odontogenic tumor was between 0.9%[23] to 12.4%,[26] and ameloblastic fibroma was 0.6%[27] to 16%.[28] The African region reported the highest frequency of

Table 2. Relative frequencies of selected odontogenic cysts

Geographic location	Years of study	Total cysts	RC cases (%)	DC cases (%)	KCOT cases (%)
Mexico[17]	10	304	118 (38.8)	108 (35.5)	57 (18.7)
Mexico[18]	21	856	342 (39.9)	283 (33.0)	184 (21.5)
France[20]	10	695	372 (53.5)	154 (22.3)	133 (19.1)
Chile[19]	28	2944	1494 (50.7)	546 (18.5)	421 (14.3)
Brazil[38]	51	2812	1772 (61.0)	735 (25.3)	208 (7.2)
UK[39]	30	7121	3724 (52.3)	1292 (18.1)	828 (11.6)
Italy[16]	19	1273	1107 (84.5)	149 (11.4)	17 (1.3)
Brazil[40]	20	680	493 (72.5)	151 (22.2)	28 (4.1)
Iran[15]	20	1227	465 (37.9)	303 (24.7)	239 (19.5)
Kuwait[41]	6	385	95 (24.7)	51 (13.2)	30 (7.8)
Total		18 297	9982 (54.6)	3772 (20.6)	2145 (11.7)

RC, radicular cyst; DC, dentigerous cyst; KCOT, keratocystic odontogenic tumor.

Table 3. Relative frequencies of selected odontogenic tumors

Geographic location	Years of study	Total number	AME cases (%)	OMY cases (%)	AOT cases (%)	AF cases (%)
Canada[22]	26	445	79 (13.5)	24 (5.4)	14 (3.1)	7 (1.6)
Mexico[25]	35	349	83 (23.7)	62 (17.7)	25 (7.1)	5 (1.4)
China[34]	42	759	445 (58.6)	64 (8.4)	63 (8.3)	14 (1.8)
Chile[27]	25	362	74 (20.4)	32 (8.8)	24 (6.6)	2 (0.6)
Estonia[28]	24	75	19 (25.3)	9 (12)	1 (1.3)	12 (16)
Nigeria[29]	33	319	201 (63)	21 (6.5)	24 (7.5)	6 (2)
Tanzania[23]	4	116	93 (80.1)	8 (7)	1 (0.9)	2 (1.7)
Nigeria[30]	19	318	233 (73)	38 (12)	9 (2.8)	10 (3)
Brazil[35]	51	340	154 (45.2)	31 (9.1)	13 (3.8)	6 (1.76)
Turkey[36]	32	527	133 (25.2)	83 (15.7)	11 (2)	8 (1.5)
India[26]	35	250	154 (61.6)	15 (6.0)	31 (12.4)	2 (0.8)
USA[24]	20	1088	127 (11.7)	24 (2.2)	19 (1.7)	17 (1.6)
Total		4948	1795 (36.3)	411 (8.3)	235 (4.7)	91 (1.8)

AME, ameloblastoma; OMY, odontogenic myxoma; AOT, adenomatoid odontogenic tumor; AF, ameloblastic fibroma.

Table 4. Relative frequencies of selected odontogenic tumors – after KCOT reclassification

Geographic location	Years of study	Total number	AME cases (%)	KCOT cases (%)	OMY cases (%)	AOT cases (%)	AF cases (%)
China[37]	52	1642	661 (40.3)	588 (35.8)	76 (4.6)	68 (4.1)	19 (1.2)
Libya[1]	17	148	33 (22.3)	52 (35.1)	5 (3.3)	4 (2.7)	2 (1.3)
Egypt[31]	15	82	34 (41.5)	16 (19.5)	7 (8.5)	3 (3.7)	2 (2.4)
China[32]	22	1309	478 (36.5)	507 (38.7)	34 (2.6)	27 (2)	13 (1)
Total		3181	1206 (37.9)	1163 (36.6)	122 (3.8)	102 (3.2)	36 (1.1)

KCOT, keratocystic odontogenic tumor; AME, ameloblastoma; OMY, odontogenic myxoma; AOT, adenomatoid odontogenic tumor; AF, ameloblastic fibroma.

ameloblastomas in the world.[23,29,30] When combining the data from all of the papers in Table 3, there were 4948 odontogenic tumors reported prior to the reclassification of KCOT. Ameloblastomas were the most frequent with 1795 (36.3%) cases. There were 411 cases of odontogenic myxoma with a frequency of 8.3%, 235 adenomatoid odontogenic tumors at 4.7%, and 91 cases of ameloblastic fibroma at a frequency of 1.8%.

Papers that published data after the reclassification of KCOT as an odontogenic tumor are presented in Table 4. The relative frequency of KCOT in these papers[1,31–33] was higher (36.6%) than was reported when KCOT was classified as an odontogenic cyst (11.7%). This is most likely due to the smaller numbers of odontogenic tumors reported as opposed to odontogenic cysts and is not a true increase in frequency.

When combining the data in Table 4 the overall frequency was 37.9% for ameloblastoma, 36.6% for KCOT, 3.8% for odontogenic myxoma, 3.2% for adenomatoid odontogenic tumor, and 1.1% for ameloblastic fibroma. These values for Table 4 are similar to those described in Table 3.

Discussion

The purpose of this investigation was to provide a comprehensive systematic review of the literature with regards to frequency of the more common odontogenic cysts and tumors. Data from the individual papers demonstrated a substantial range in frequency for the various pathoses presented.

By fulfilling the inclusion criteria, the papers selected were found to be the most acceptable in the literature. The most appropriate method available for analyzing the data was to combine the results from the various papers. By pooling the data, the results could be employed across the world when healthcare professionals are discussing the possibility that their patient has one of these lesions.

The specific odontogenic cysts and tumors were chosen for this review as they were the most common lesions across the literature. This review has found that odontogenic cysts are 2.25 times more frequent than odontogenic tumors.

There were several different odontogenic cysts that were diagnosed less frequently including: calcifying odontogenic cyst, lateral periodontal cyst, eruption cyst, botryoid odontogenic cyst, and residual cyst. Collectively, these other cysts made up less than 10% of the total number of cysts.[15–20] Not all studies reviewed had these other variants present, therefore no comparison could be made. It was not surprising that radicular cysts were the most frequent cystic lesions, as these cysts can be associated with any tooth in the mouth following dental pulp necrosis, which can lead to radicular cyst formation.

Most of the papers that reported odontogenic tumors had sporadic cases of calcifying epithelial odontogenic tumors, cementoblastomas, clear cell odontogenic tumors, and squamous odontogenic tumors. These less frequent tumors were only a small proportion of the overall total.[1,22–24,26–32,34–37] Again each of the papers reviewed had variations with these less common tumors, therefore comparison between them was not suitable.

As odontogenic cysts and tumors are not notifiable to any cancer or diseases registry, it is difficult to ascertain whether any of the data published are truly accurate. To illustrate the point, odontomas are a benign hamartomatous conglomerate of dental tissue, where there is controversy about this lesion being a tumor at all. Odontomas are reported in a number of papers presenting data on odontogenic tumors with a frequency of up to 45.8%.[22] Some practitioners may not even refer this lesion for histopathological review; therefore it would be difficult to gauge the true number of odontomas in a population. The same could be considered for any other odontogenic cyst or tumor to some degree. In particular the radicular cyst is probably underreported as clinicians may rely on their clinical judgment to diagnose the pathology while extracting a tooth.

The reclassification of KCOT from a cyst to a tumor by the WHO in 2005[13] appears to have minimal influence on the frequency of other odontogenic tumors. The overall frequency of KCOT has risen from 11.7% when it was classified as an odontogenic cyst, to 36.6% since classification as an odontogenic tumor, appearing as a threefold increase in the frequency of KCOTs.

Gaitan-Cepeda *et al.*[33] reviewed the impact of this reclassification of KCOT as an odontogenic tumor for two time periods (1981–2004 and 2005–2008). It was noted that the prevalence of odontogenic tumors in histopathology accessions increased from 2.6% (1981–2004) to 5% (2005–2008). Interestingly, the prevalence of odontogenic cysts was similar in both time periods; 8.7% (1981–2004) and 7.7% (2005–2008).[33] For the other odontogenic tumors, the overall frequencies have been relatively stable when comparing Tables 3 and 4. For example, ameloblastoma was 36.3% prior to KCOT inclusion and 37.9% after KCOT inclusion.

The overall frequencies provided in this systematic review are applicable to populations around the world presenting average values. Papers with African data certainly present higher frequencies for ameloblastomas. However, Simon *et al.*[23] felt that the main reason for the seemingly high frequency was due to patients presenting to hospital only when they had debilitating symptoms.

Conclusion

This systematic review has analyzed the literature to provide an overall frequency of the most common odontogenic cysts and tumors throughout the world. It is not surprising that radicular cysts are the most frequent odontogenic cyst, followed by the dentigerous cyst and KCOT (when classified as a cyst). Ameloblastomas are the most frequent odontogenic tumor, followed by KCOT (when classified as a tumour), odontogenic myxoma, adenomatoid odontogenic tumor, and ameloblastic fibroma. The reclassification of KCOT to an odontogenic tumor has made it the second most frequent odontogenic tumor, increasing the reported numbers in this group, but this has not significantly decreased the frequencies of the other odontogenic tumors.

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Conflicts of interest

The authors report no conflicts of interest.

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